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Case Report

Effect of the Lee Silverman Voice Treatment on Hypophonic Dysarthria from Post-stroke Parkinsonism: A Case Report

Jong Mi Park, M.D., Yong Wook Kim, M.D., Ph.D., Su Jung Kim, Ph.D.[1] and Soojin Choi, M.D.

Department and Research Institute of Rehabilitation Medicine, Yonsei University College of Medicine, Seoul, Republic of Korea
[1]Speech-language and Psychological Therapy, Department of Rehabilitation Medicine, Severance Hospital, Yonsei University College of Medicine, Seoul, Republic of Korea

조음장애로 진단된 뇌졸중 후 파킨슨증의 리실버만 음성 치료 효과: 증례보고

박종미 · 김용욱 · 김수정[1] · 최수진

연세대학교 의과대학 재활의학교실 및 재활의학연구소, [1]연세대학교 의과대학 세브란스 재활병원 재활2팀 언어 · 심리치료실

Abstract

Post-stroke parkinsonism usually presents with bradykinesia, resting tremor, and movement disabilities and reportedly responds poorly to rehabilitative and pharmacological treatment in contrast to Parkinson's disease. However, we encountered a patient with subarachnoid hemorrhage caused by a ruptured anterior communicating artery aneurysm who developed parkinsonism, which manifested with hypokinetic hypophonic dysarthria and masked facies. Brain ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography revealed decreased glucose metabolism in the bilateral basal ganglia. He underwent 18 sessions of Lee Silverman Voice Treatment (LSVT) for 60 min, once daily, and he gradually increased the previously prescribed doses of levodopa and benserazide to 200 mg and 50 mg, respectively, three times a day. The patient's dysarthria improved from moderate to mild dysarthria. His masked facies also improved remarkably 6 weeks after admission. Along with levodopa administration, LSVT could be suggested as an effective treatment tool for hypophonic dysarthria due to post-stroke parkinsonism.

Key Words

Post-stroke parkinsonism, Hypophonic dysarthria, Lee Silverman Voice Treatment

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Corresponding author : Soojin Choi, M.D.
Department of Rehabilitation Medicine, Yonsei University College of Medicine, 50-1 Yonsei-ro, Seodaemun-gu, Seoul 03722, Republic of Korea
Tel : 82 2 2228 3722
Fax : 82 2 2227 8341
e-mail : sjc8070@yuhs.ac

Introduction

While idiopathic Parkinson's disease (IPD) is a neurodegenerative disease that occurs in the aging brain, many other causes of parkinsonism, such as vascular parkinsonism or post-stroke parkinsonism, also show correlation with advancing age.[1] Post-stroke parkinsonism, which reportedly occurs in 1-4% of cases after stroke,[2] is mainly characterized by gait disturbances (broad-based, shuffling, or freezing gait), postural instability, tremor,

and bradykinesia. However, we encountered a patient with a ruptured aneurysm and subarachnoid hemorrhage who developed parkinsonism and presented with marked dysarthria and masked facies instead of the typical features of post-stroke parkinsonism.

The Lee Silverman Voice Treatment (LSVT), developed by Ramig et al. in 1987, is an intensive voice treatment. It includes tasks that require patients to speak aloud, which maximizes their voice intensity and breathing ability and provides an effective speech therapy for patients with speech disorders related to IPD.[3] However, few studies have focused on its efficacy on secondary parkinsonism. Our patient showed marked improvement in hypokinetic hypophonic dysarthria and masked facies after LSVT. Although most previous case reports have stated that secondary parkinsonism does not respond well to levodopa treatment,[4] the patient reported herein showed considerable improvement through LSVT and levodopa administration. Therefore, we report a patient with unusual symptoms of post-stroke parkinsonism that significantly improved through intensive speech treatment.

Case Report

A 48-year-old Asian man had a medical history of hypertension, diabetes mellitus, and dyslipidemia for 3 years, all of which were controlled with oral medications. He had no history of parkinsonism before the onset of stroke. He also had no history of other medications such as atypical antipsychotic drugs associated with drug-induced parkinsonism. He was diagnosed with an anterior communicating artery aneurysm measuring 1 cm × 0.8 cm and subarachnoid hemorrhage and underwent embolization at another hospital the same day. He was transferred to our hospital for rehabilitative treatment 3 months after the onset of stroke.

On the day of hospitalization, his speech was unintelligible owing to the monotony of his words and low intensity of his voice. Moreover, his hypomimia made it difficult to identify his emotions. The patient's family often misconstrued his condition as depression. His muscle strength was relatively well preserved upon initial assessment at our hospital: the Medical Research Council Scale for muscle strength was 5 on the right side and 4 on the left side. Gait disturbances, postural instability, tremors, and bradykinesia were not observed.

The patient was subsequently diagnosed with post-stroke parkinsonism based on the above-mentioned symptoms. He was assessed using the motor examination segment of the Unified Parkinson's Disease Rating Scale-Part III (UPDRS-III). His initial motor examination score on the UPDRS-III was 7 out of 56; the speech and facial expression scores were 3 and 4, respectively. We also evaluated his vocalization and speech using the LSVT LOUD protocol (see Supplemental appendix A for details).[5,6] His initial maximum phonation time for the sustained phonation of /a/ was 4.22 s, with a sound pressure level of 76 dB. The initial sound pressure levels while reading two words were 75.37 dB and 71 dB while reading a paragraph. The highest pitch for the phonation of /a/ was 210 Hz, and the lowest pitch was 133 Hz. The pitch range was narrow and monotonous at 77 Hz. Individual speech sound could be heard, but he exhibited a low, rapid, murmur type of speech. His overall speech intelligibility was very poor, and it was difficult for others to understand him. He was then diagnosed with hypokinetic hypophonic dysarthria (moderate to severe), a communication disorder. Subsequently, speech therapy was prescribed with the aim of improving his phonation and dysarthria to enhance his speech intelligibility at short word levels.

Initial brain computed tomography performed at onset of stroke showed no other abnormalities except for the subarachnoid hemorrhage and intracerebral hemorrhage in the frontal region (Fig. 1A). Brain computed tomography performed 3 months after injury showed a low-attenuating lesion in the left frontal lobe without hydrocephalus (Fig. 1B). A functional imaging test was also performed to determine the cause of the patient's persistent symptoms. Brain ^{18}F-fluorodeoxyglucose positron emission

tomography/computed tomography (^{18}F-FDG-PET/CT) performed 1 week after admission revealed decreased glucose metabolism in the bilateral basal ganglia and multiple ischemic regions in the bilateral cerebral cortex (Fig. 1C).

Dysarthria and hypomimia occurred with the onset of stroke. Two months after the initial symptom development, he was administered 100 mg levodopa and 25 mg benserazide hydrochloride once a day at the previous hospital for one month. However, he did not show any improvement during this time. He was then admitted to our hospital 3 months after the initial onset, and we gradually increased the prescribed doses of the levodopa/benserazide combination. Two days after his admission, the levodopa/benserazide was increased to 50 mg/12.5 mg three times a day, and after 3 days to 150 mg/37.5 mg three times a day, and after another 3 days to a final dose of 200 mg/50 mg three times a day. However, the patient still did not show significant improvement of dysarthria and hypomimia. Since the patient did not show significant improvement with pharmacologic treatment, speech therapy was started on the 7th day from the day levodopa/benserazide was increased to the final dose. Following the LSVT LOUD protocol, LSVT was performed by a qualified and experienced speech therapist with an LSVT LOUD certification for the treatment of parkinsonian dysarthria. The patient received a total of 18 sessions of LSVT lasting 60 min each day for 4 weeks. He also received comprehensive rehabilitation therapy, including physical therapy twice a day, and occupational therapy once a day for 30 min each.

The follow-up speech evaluation was performed 4 weeks from the initial evaluation. He was medically stable,

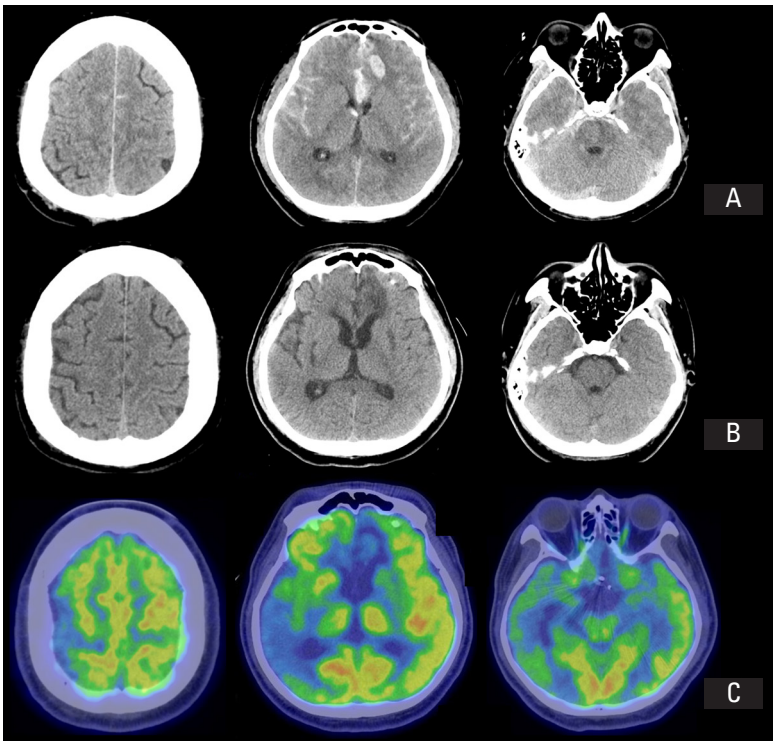

Fig. 1. (A) Initial brain computed tomography showing subarachnoid hemorrhage and intracerebral hemorrhage in the left frontal lobe owing to the ruptured aneurysm. (B) Brain computed tomography obtained 3 months after stroke showing decreased attenuation in the left frontal lobe without hydrocephalus. (C) Brain 18F-fluorodeoxyglucose positron emission tomography-computed tomography images revealing decreased glucose metabolism in bilateral basal ganglia, left frontal lobe and bilateral cerebral cortices.

and there was no difference in his medical status during the initial versus the follow-up evaluation. As primary outcomes, the maximum phonation time, pitch range, and sound pressure levels, which were the values that have shown improvement on LSVT treatment for Parkinson's disease in previous studies, were measured.[7-9] The patient's mean maximum phonation time improved from 4.22 s to 9.48 s after the cessation of the LSVT sessions (Fig. 2A). The sound pressure level improved from 76 dB to 88.2 dB when the patient consistently produced the /a/ sounds (Fig. 2B). The highest pitch during the phonation of /a/ improved from 210 Hz to 229.66 Hz. The lowest pitch during the phonation of /a/ improved from 133 Hz to 104 Hz. The pitch range increased from 77 Hz to 133.67 Hz, indicating an improvement in pitch control (Fig. 3A). Sound pressure levels improved when the patient read two succeeding

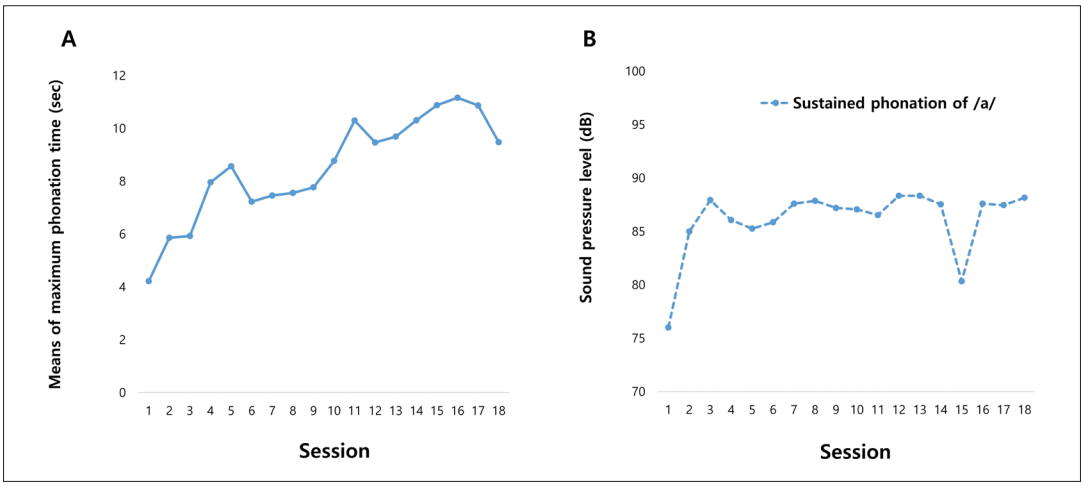

Fig. 2. (A) The mean of the maximum phonation time (in seconds) improved after the Lee Silverman Voice Treatment. (B) Sound pressure levels (dB) during the sustained phonation of /a/ also improved with the Lee Silverman Voice Treatment.

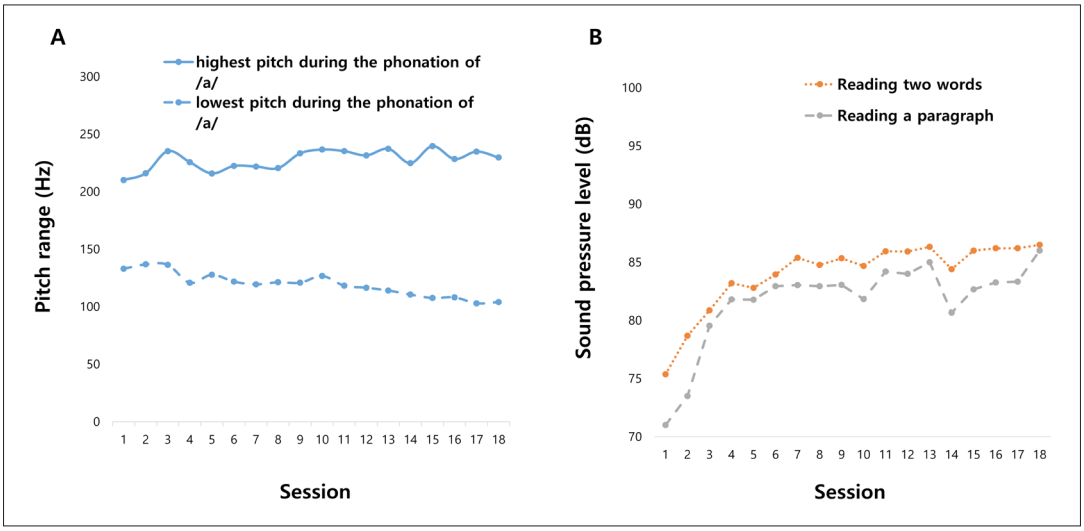

Fig. 3. (A) The sound pitch range during the phonation of /a/ widened, thus improving the patient's pitch control. (B) Sound pressure levels (in dB) while the patient read two succeeding words or a full paragraph improved, allowing the patient a louder and clearer speech.

words to full paragraphs, indicating an improvement in his speech intelligibility (Fig. 3B). His moderate dysarthria was improved to a mild degree, and this improvement was reflected in his speech clarity. He exhibited mild dysarthria at sentence-level speech intelligibility, and only occasional unnatural intonation and syllable clustering were observed. Improvements in dysarthria made social communication possible.

His masked facies also improved. On the first day of hospitalization, he had mask-like facial expressions and decreased blinking. The expression of his mouth improved gradually, and 6 weeks following his admission, he was able to smile using the zygomatic, orbicularis oris, and orbicularis oculi muscles (Fig. 4). His UPDRS-III score decreased from 7 out of 56 to 2 on the day of his discharge. His speech and facial expressions improved greatly to 1 point each. The overall score improved by approximately 71.4% (Table 1).

Fig. 4. Facial expression of the patient at (A) day 1 of hospitalization, (B) four weeks after hospitalization, and (C) six weeks after hospitalization.

Table 1. Unified Parkinson's Disease Rating Scale-Part III (UPDRS-III) (Motor Examination) Scores Upon Initial Assessment and Discharge

UPDRS-III subscales	Initial score	Final score at discharge
Speech	3	1
Facial expression	4	1
Tremor at rest	0	0
Action or postural tremor of hands	0	0
Rigidity	0	0
Finger taps	0	0
Hand movements	0	0
Rapid alternating movements of hands	0	0
Leg agility	0	0
Arising from chair	0	0
Posture	0	0
Gait	0	0
Postural stability	0	0
Body bradykinesia and hypokinesia	0	0
Total score	7	2

Discussion

It is well known that IPD mainly affects the elderly, with two-thirds of the patients being over the age of 70 years.[10] However, there are many other major causes of parkinsonism in the geriatric population, and the prevalence of parkinsonism increases with advancing age.[11] In addition, the proportion of parkinsonism not due to IPD also increases with age.[10] Therefore, effective treatment for secondary parkinsonism will be helpful in managing the elderly with various symptoms of parkinsonism.

In our patient, post-stroke parkinsonism with features of hypomimia and dysarthria improved significantly with LSVT and levodopa administration. Patients with post-stroke parkinsonism generally present with bradykinesia, resting tremor, muscular rigidity, and gait impairment.[12] Secondary parkinsonism, including post-stroke parkinsonism, has been known to be less responsive to levodopa treatment. Our case is rare and worth reporting since the patient's principal presenting symptoms included dysarthria and hypomimia, which are considered axial symptoms and are uncommon in post-stroke parkinsonism. Moreover, the patient responded well to pharmacological therapy along with intensive speech therapy, in contrast to the results of previous studies.[4]

Our therapeutic approach was focused on speech rehabilitation for alleviating the symptoms of dysarthria. More than 75% of patients with Parkinson's disease have language and speech abnormalities.[3] Sound reduction, lack of expression, difficulty in breathing and hoarseness affect speech clarity, language, and communication. These abnormalities are caused by inadequate vocal fold adduction, reduced laryngeal muscle activation or synergy, muscle atrophy or fatigue, asymmetric vocal fold tension or movements, and stiffness or rigidity of the vocal folds and respiratory muscles.[13] Our patient initially exhibited mild hoarseness in his voice and spoke in a low murmur. Moreover, resting tremors were frequently observed on the lips and tongue, limiting the speed and range of movement of his entire speech and articulation system. To date, the LSVT program developed by Ramig et al. is the most effective treatment for Parkinson's disease-related speech disorders. It involves an intensive structured program: each session consists of a set of simple tasks designed to maximize the patient's voice and breathing abilities. The therapist instructs and constantly stimulates the patient to speak aloud with emphasis on constant conversation and various speech activities.[14] The LSVT LOUD protocol was also shown to be the most effective when it was performed for 4 consecutive days per week (16 sessions per month) for 4 weeks.[5] LSVT was performed according to the established protocol, and the patient's dysarthria improved from moderate to mild dysarthria. His voice length, voice intensity, tone control, and speech intelligibility improved after the LSVT.

The patient's masked facies also improved after LSVT. The reduction in spontaneous expression without facial paralysis in patients with Parkinson's disease is called "masked facies" or "hypomimia." Hypomimia has been reported in up to 70% of patients with Parkinson's disease.[15] Facial mobility was reportedly reduced in patients with Parkinson's disease along with considerable retardation in the time required to reach peak expression (bradykinesia) compared to that in a control group.[16] Therefore, these patients smile less frequently and with a lower intensity. According to previous studies, LSVT is known to be effective for hypomimia observed in idiopathic parkinsonism.[17] However, little is known about the presentation of masked facies in post-stroke parkinsonism. This case report supports that LSVT is also helpful in restoring various facial expressions in patients with dysarthria and hypomimia related to post-stroke parkinsonism. Levodopa is effective for treating the motor and non-motor symptoms of IPD but shows poor therapeutic efficacy for secondary parkinsonism.[18] However our patient responded well to increased dosage of levodopa in combination with speech therapy.

Our case report has some limitations. First, our patient was diagnosed with post-stroke parkinsonism through clinical symptoms and brain ^{18}F-FDG-PET/CT results, but

other functional imaging techniques such as Dopa-PET or single-photon emission computed tomography (SPECT) were not performed. Dopamine transporter imaging, which aids in assessing the integrity of the dopaminergic pathway, is abnormal in IPD and other Parkinson-plus syndromes, and a normal scan can suggest an alternative diagnosis such as vascular parkinsonism or drug-induced parkinsonism.[19,20] The pathological background of vascular parkinsonism is poorly understood, and there are no generally accepted critera.[21,22] However, an updated diagnostic approach in 2018 subdivides vascular parkinsonism into three subdivisions: acute/subacute post-stroke vascular parkinsonism, insidious vascular parkinsonism, and mixed neurodegenerative parkinsonism and cerebrovascular disease.[23] Our patient can be assumed to be included in the acute/subacute post-stroke vascular parkinsonism due to the non-progressive clinical condition with timely relation to the occurrence of the stroke.[23] However, the use of dopaminergic imaging may still have helped for more accurate diagnosis.[19,23,24]

The second limitation is that, since LSVT and levodopa administration were combined, it is difficult to conclude that the improvement of symptoms is a result of LSVT alone. However, the patient had already been taking levodopa and benserazide for 1 month at the time of admission but had no improvement in symptoms. Considering the effective period of levodopa and benserazide, which is 1-3 weeks,[25] and since he did not show any improvement even with increased dosage, it is highly likely that LSVT for 4 weeks mainly led to the improvement in his symptoms. Nonetheless, the effect of LSVT and levodopa combination therapy cannot be excluded.

This case report suggests that LSVT along with levodopa administration can be effective for managing the axial symptoms of post-stoke parkinsonism. Further studies are needed to determine whether LSVT monotherapy is effective without levodopa in post-stroke parkinsonism. This report will be helpful for the management of secondary parkinsonism, especially those with features of dysarthria and masked facies.

REFERENCES

1. Demirkiran M, Bozdemir H, Sarica Y. Vascular parkinsonism: a distinct, heterogeneous clinical entity. Acta neurologica scandinavica 2001;104:63-67
2. Handley A, Medcalf P, Hellier K, Dutta D. Movement disorders after stroke. Age and Ageing 2009;38:260-266
3. Ramig LO, Sapir S, Countryman S, Pawlas AA, O'Brien C, Hoehn M, et al. Intensive voice treatment (LSVT) for patients with Parkinson's disease: a 2 year follow up. J Neurol Neurosurg Psychiatry 2001;71:493-498
4. Korczyn AD. Vascular parkinsonism—characteristics, pathogenesis and treatment. Nature Reviews Neurology 2015;11:319
5. Fox C, Ebersbach G, Ramig L, Sapir SJPsd. LSVT LOUD and LSVT BIG: behavioral treatment programs for speech and body movement in Parkinson disease 2012;2012
6. Fox C, Ebersbach G, Ramig L, Sapir S. LSVT LOUD and LSVT BIG: Behavioral Treatment Programs for Speech and Body Movement in Parkinson Disease. Parkinsons Dis 2012;2012:391946
7. Byeon H. The combined effects of lee silverman voice treatment and abdominal respiration on the maximum phonation time in parkinson's disease. International Journal of Engineering and Technology (UAE) 2018;7:86-89
8. Whitehill TL, Wong LLN. Effect of intensive voice treatment on tone-language speakers with Parkinson's disease. Clinical linguistics & phonetics 2007;21:919-925
9. Ramig LO, Sapir S, Fox C, Countryman S. Changes in vocal loudness following intensive voice treatment (LSVT®) in individuals with Parkinson's disease: A comparison with untreated patients and normal age-matched controls. Movement disorders: official journal of the Movement Disorder Society 2001;16:79-83
10. Meara J, Koller WC. Parkinson's Disease and Parkinsonism in the Elderly: Cambridge University Press; 2000
11. Barbosa MT, Caramelli P, Maia DP, Cunningham MCQ, Guerra HL, Lima-Costa MF, et al. Parkinsonism and Parkinson's disease in the elderly: A community-based

survey in Brazil (the Bambuí study). Movement Disorders 2006;21:800-808

12. Fitzgerald PM, Jankovic JJMdojotMDS. Lower body parkinsonism: evidence for vascular etiology 1989;4:249-260

13. Baker KK, Ramig LO, Luschei ES, Smith ME. Thyroarytenoid muscle activity associated with hypophonia in Parkinson disease and aging. Neurology 1998;51:1592-1598

14. Dromey C, Ramig LO. Intentional changes in sound pressure level and rate: Their impact on measures of respiration, phonation, and articulation. Journal of Speech, Language, and Hearing Research 1998;41:1003-1018

15. Rinn WE. The neuropsychology of facial expression: a review of the neurological and psychological mechanisms for producing facial expressions. Psychological bulletin 1984;95:52

16. Bowers D, Miller K, Bosch W, Gokcay D, Pedraza O, Springer U, et al. Faces of emotion in Parkinsons disease: micro-expressivity and bradykinesia during voluntary facial expressions. J Int Neuropsychol Soc 2006;12:765-773

17. Dumer AI, Oster H, McCabe D, Rabin LA, Spielman JL, Ramig LO, et al. Effects of the Lee Silverman Voice Treatment (LSVT® LOUD) on hypomimia in Parkinson's disease 2014;20:302-312

18. Ricciardi L, De Angelis A, Marsili L, Faiman I, Pradhan P, Pereira E, et al. Hypomimia in Parkinson's disease: an axial sign responsive to levodopa 2020

19. Thanvi B, Lo N, Robinson T. Vascular parkinsonism—an important cause of parkinsonism in older people. Age and ageing 2005;34:114-119

20. Eshuis SA, Jager PL, Maguire RP, Jonkman S, Dierckx RA, Leenders KL. Direct comparison of FP-CIT SPECT and F-DOPA PET in patients with Parkinson's disease and healthy controls. Eur J Nucl Med Mol Imaging 2009;36:454-462

21. Kalra S, Grosset DG, Benamer HT. Differentiating vascular parkinsonism from idiopathic Parkinson's disease: a systematic review. Movement Disorders 2010;25:149-156

22. Vizcarra JA, Lang AE, Sethi KD, Espay AJ. Vascular P arkinsonism: D econstructing a S yndrome. Movement Disorders 2015;30:886-894

23. Rektor I, Bohnen NI, Korczyn AD, Gryb V, Kumar H, Kramberger MG, et al. An updated diagnostic approach to subtype definition of vascular parkinsonism–Recommendations from an expert working group. Parkinsonism & related disorders 2018;49:9-16

24. Thobois S, Prange S, Scheiber C, Broussolle E. What a neurologist should know about PET and SPECT functional imaging for parkinsonism: a practical perspective. Parkinsonism & related disorders 2019;59:93-100

25. Numbers L, Cost BNJJoN, Neurosurgery,, Psychiatry. Madopar 1983

Supplemental Appendix A. LSVT LOUD Protocol[6]

Target
Increased movement amplitude directed predominately to respiratory/laryngeal systems
Intensity: standardized
Dosage: 4 consecutive days a week for 4 weeks (16 sessions in one month) Repetitions: minimum 15 repetitions/task Effort: push for maximum patient-perceived effort each day (8 or 9 on scale of 1–10 with 10 being the most)
Daily exercises
First half of the treatment session (30 min.) Task 1: Maximum Sustained Movements 15 reps: sustain “ah” in Loud good quality voice as long as possible Task 2: Directional Movements 15 reps each: say “ah” in Loud good quality voice going high in pitch; 15 reps each: say “ah” in Loud good quality voice going low in pitch Task 3: Functional Phrases Patient self-identifies 10 phrases or sentences he/she says daily in functional living (e.g., “Good morning”) 5 reps of the list of 10 phrases. “Read phrases using same effort/loudness as you did during the long “ah”
Hierarchy
Second half of the treatment session (30 min) (i) Designed to train rescaled amplitude/effort of movement achieved in daily exercises and functional phrases into in context specific and variable speaking activities (ii) Tasks increase complexity across weeks (Words-phrases-sentences-reading-conversation) and can be tailored to each patient’s goals and interests (e.g., golf versus cooking) (iii) Tasks progress in difficulty by increasing duration (maintain LOUD for longer periods of time) amplitude (loudness, within normal limits), and complexity of tasks (dual processing, background noise, and attentional distracters)
Shaping techniques
Goal: train vocal loudness that is healthy and good quality (i.e., no unwanted vocal strain or excessive vocal fold closure) Technique: shape the quality and voice loudness through use of modeling or tactile/visual cues. “Watch me and do what I do.” Minimal cognitive loading: behavior is not achieved through extensive instructions or explanations, which are often too complex for patients to generalize outside of the treatment room, but rather the patient is trained through modeling
Sensory recalibration
Treatment: focus attention on how it feels and sounds to talk LOUD Carryover activities: start day one; daily assignments (treatment and nontreatment days); Loud good quality voice in real-life situations; (i) difficulty of the assignment matches the level of the hierarchy where the person is working; (ii) make patient accountable and probe for comments from patient that people in their daily living have said, such as, “I can hear you better” Homework practice: start day one: daily assignments to practice at home (Daily Exercises and Hierarchy Exercises); treatment days (one other time for 5–10 minutes); nontreatment days (two times for 10–15 minutes); homework book provided and patient made accountable